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Strategies for Conducting Research on Language in Autism

Helen Tager-Flusberg[1]

Several different methodological approaches that have been used in studying language in children with autism are outlined. In classic studies, children with autism are compared to comparison groups typically matched on age, IQ, or mental age in order to identify which aspects of language are uniquely impaired in autism. Several methodological problems are noted with this approach including (a) heterogeneity of the autism population, (b) mental retardation, (c) developmental change with age, and (d) sample size and ascertainment. An alternative strategy is suggested which focuses on identifying the complex expression of the language phenotype in autism across the full range of the syndrome. This approach explores within-group individual differences in language functioning, and recently identified distinct language phenotypic subgroups within the autism population that are relevant to understanding the underlying genetic and neurobiological etiology of autism.

KEY WORDS: Autism; genetics; language; phenotype; subtypes.

INTRODUCTION

Classic Studies of Language in Autism

Impairments in language are among the core features of autism (APA, 1994). Although Kanner (1943) did not consider language central to the description of the new syndrome that he introduced, he later outlined some of the unusual clinical features that distinguished the language of children with autism from children with other disorders (Kanner, 1946). Research on language conducted over the past 50 years has primarily been concerned with this question: What are the unique and universal features that define the abnormal language characteristics of autism?

Following up on Kanner's (1946) observations, early clinical studies were focused on describing the abnormal language used by children with autism, which included atypical intonation and vocal quality, idiosyncratic use of words and stereotyped phrases, echolalia, and pronoun reversal (e.g., Cunningham, 1966; Goldfarb, Braunstein, & Lorge, 1956; Pronovost, Wakstein, & Wakstein, 1966; Shapiro & Fish, 1969). The majority of these published reports were neither methodologically systematic nor developmentally informed as they relied mostly on poorly defined clinical samples or single case histories. Nevertheless, the features that were highlighted in these studies remain important for diagnosing autism spectrum disorders, and they are included in most diagnostic classification systems and instruments (e.g., APA, 1994; Krug, Arick, & Almond, 1980; Lord *et al.,* 1994, 2000; Schopler, Reichler, De Vellis, & Daly, 1980; WHO, 1993).

By the mid 1970s, more carefully designed studies were conducted that were framed in the methodology used by developmental psycholinguists. Because the goals of these studies were to identify unique features of language in autism—features that distinguish between autism and other populations—they generally included *matched* groups of children with autism and some other comparison group or groups. Comparison groups included children with other language disorders, mental retardation, or typically developing children (e.g., Bartak, Rutter, & Cox, 1975, 1977; Bartoclucci,

[1] Correspondence should be addressed to Helen Tager-Flusberg, Ph.D., Department of Anatomy and Neurobiology, Boston University School of Medicine, 715 Albany Street L-814, Boston, MA 02118; e-mail: htagerf@bu.edu

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Pierce, Streiner, & Eppel, 1976; Pierce & Bartoclucci, 1977; Tager-Flusberg, 1981a, 1985) in designs that were introduced in the seminal cross-sectional experimental studies conducted by Hermelin and O'Connor (1970). In most studies on language in autism, groups were matched on IQ or mental age in an effort to control for the influence of mental retardation on language. During this period, studies were primarily focused on receptive or expressive phonological, syntactic, morphological, or semantic ability and addressed the question of whether children with autism differed from matched control groups on these aspects of language. The general conclusions were that they did not (Tager-Flusberg, 1981b).

Later studies on pragmatic functioning in autism followed this same pattern. Initially, clinical descriptions were published (e.g., Baltaxe, 1977) followed by more systematic empirical investigations that included control groups matched on IQ, mental age, or, more often, other aspects of language ability such as vocabulary or syntax (e.g., Capps, Kehres, & Sigman, 1998; Loveland, Landry, Hughes, Hall, & McEvoy, 1988; Norbury & Bishop, 2002; Surian, Baron-Cohen, & Van der Lely, 1996). In other studies, unique patterns of language organization among children with autism were explored in relation to dissociations between pragmatic and syntactic or lexical components of language functioning in contrast to more synchronous patterns found among control groups (e.g., Tager-Flusberg & Anderson, 1991; Tager-Flusberg, 1994). The consistent picture that emerged from these studies was that pragmatic aspects of language were specifically and universally impaired in children with autism relative to other children and to other aspects of language (Lord & Paul, 1997; Tager-Flusberg, 2000).

Overall, research in which the language abilities of children with autism was compared to language abilities in other matched groups of children yielded important findings about core and defining deficits in the domain of pragmatics. Nevertheless, many, if not all of these studies were plagued by methodological problems, as are studies investigating other areas of cognitive or behavioral functioning in autism.

METHODOLOGICAL PROBLEMS

There are numerous methodological concerns that can be identified in the literature on language in autism. Because diagnostic criteria changed significantly over the past five decades, and there was, until recently, a dearth of objective methods for researchers to use to define autism or autism spectrum disorders, it is doubtful that across studies the same criteria were used for including children under the diagnostic label of "autism." These diagnostic issues began to be overcome in recent studies through reaching a consensus in both the clinical and research communities about how to define autism and associated disorders (APA, 1994; WHO, 1993) and through the introduction of gold standard methods for achieving reliable diagnoses using the Autism Diagnostic Interview–Revised version (ADI-R; Lord *et al.,* 1994) and the Autism Diagnostic Observation Schedule (ADOS; Lord *et al.,* 2000). Despite these advances in diagnosis, there are other methodological problems that undermine many cross-sectional studies in this field, especially language studies in which groups of persons with autism are matched to comparison participants.

Heterogeneity of the Population

By definition, autism is a complex disorder that is heterogeneous in the expression of both defining and associated symptoms. This heterogeneity poses a significant challenge in the selection of participants for research. Typically, most researchers do not assess participants on all the symptoms that are associated with the syndrome of autism. For example, we may include some participants with associated anxiety and mood disorders and some without. Co-morbid psychiatric problems can potentially influence performance on language tasks, yet we generally do not control for these issues in selecting either autism participants or participants in matched groups. In the domain of language, there is enormous heterogeneity, ranging from children with little to no functional language to those with scores on standardized measures that are well above the average range. This kind of variance may easily mask potentially significant group differences of certain features of language in studies that follow the classic matched group design.

Mental Retardation

The majority of children with autism also have mental retardation, which may vary from borderline to profound. At the same time, about one-quarter or more of the population do not have mental retardation, and some children with autism score in the superior range on standard intelligence tests. Both children with and without mental retardation are included in many studies, even though intelligence is significantly correlated with language ability. Although this factor is often controlled by matching comparison groups on mental age

or IQ, matching is often defined as nonsignificant differences on some parametric statistic, even though the range of scores is not the same for the persons with autism and the comparison participants.

Developmental Change with Age

Autism is a developmental disorder. Even the most impaired preschoolers with autism can show significant developmental advances in language as they grow older, just as nonautistic preschoolers do. Yet many studies include participants that vary significantly in age, and these key developmental issues are ignored. The matching of groups that vary widely in age does not resolve the problem; rather, it is simply extended to the comparison groups. By including children of widely differing ages, there is an implicit assumption that performance on the key language task or measure is not influenced by developmental change. This assumption is patently not true for almost any aspect of language, though it may be ameliorated if standard score measures, which are adjusted for age, are the key dependent variables.

Sample Size and Ascertainment

Autism is a relatively rare disorder and it is often difficult for researchers to obtain a large number of participants for their studies. Most participants are identified through school or clinic referrals, which may bias the ascertainment of samples for language studies to those children that have more severe language or communicative impairments. The typical autism language study includes relatively small numbers of participants and we do not carefully assess whether studies have sufficient power to detect differences between the matched groups that are included.

These four problems plague the majority of empirical studies that address the question of whether children with autism have specific deficits in language by comparing matched groups on language measures. Furthermore, these problems are interrelated as heterogeneity in autism includes many associated symptoms as well as IQ, language, and age. The inclusion of groups of children with autism that vary widely on all these dimensions requires large numbers of participants if studies are to meet minimal power requirements. At the same time, including a narrow range of children (e.g., only those with normal intelligence) does not allow researchers to investigate whether children with retardation may have certain language deficits that are not found among nonretarded children with autism.

INVESTIGATING VARIATION IN LANGUAGE IN AUTISM

The emphasis of much of the autism research of the past several decades on discovering the unique and universal features of language abnormalities led to the neglect of a different, but equally significant research question that needs to be addressed: What is the language phenotype of autism? The scope of this question is significantly broader than the earlier focus on identifying only those aspects of language impairment that distinguish autism from other populations. Moreover, the full range of language abilities and impairments in autism, which together make up the phenotypic expression of this component of the disorder, cannot easily be investigated using the same methods used in earlier research. Cross-sectional matched group designs will not be useful for studying variation in language among persons with autism; instead, a within-group individual approach may be better suited to address this kind of research question.

Recently, we embarked on a series of studies that explicitly focused on exploring the heterogeneity of language abilities among children with autism (Kjelgaard & Tager-Flusberg, 2001; Tager-Flusberg, 2003) with a within-group design. The first study (Kjelgaard & Tager-Flusberg, 2001) included 89 children between the ages of 4 and 14, all of whom were diagnosed with autism using the ADI-R and ADOS. We recruited children who had at least acquired phrase speech, thus curtailing our investigation to investigating heterogeneity among *verbal* children with autism. The sample varied widely in IQ, with scores on the Differential Ability Scales ranging from 25 to 141. Each child was administered a battery of standardized language tests testing articulation skills, receptive and expressive vocabulary, nonsense word repetition skills, and higher-order receptive and expressive syntax and semantics. About half the group was not able to complete testing on the more advanced language tests because of their limited cognitive and linguistic abilities.

As expected, we found a wide range of performance using standard scores on each of the language measures. Some children scored at or above the mean, whereas others scored well below the mean, in the significantly impaired range. More relevant to the issue of phenotypes, these analyses identified different language subtypes among the children. One subtype, representing about one-quarter of the sample, scored within the normal range across all the tests that were administered. While the majority of children in this "normal" subtype

also had IQ scores in the normal range, some were mentally retarded, indicating that language subtypes are not fully determined by IQ. A second group of children, about half the sample, scored more than one or two standard deviations below the mean across most of the language tests, representing the "impaired" language subtype. This impaired subtype included children with both normal IQ scores and children with mental retardation. The remaining children scored in the borderline or normal range across the different tests and did not fit a consistent subtype pattern. The profile of performance for the children in the "impaired" subtype across the different tests was very striking. These children had unimpaired articulation skills, and in general scored lower on the higher order syntactic and semantic measures than on the vocabulary or nonword repetition tests. This profile is the same as has been found for children with specific language impairment (SLI), a different language disorder (Tomblin & Zhang, 1999).

In follow-up studies, we investigated whether this subtype of children with "impaired" language displays the same language deficits as children with SLI (Tager-Flusberg, in press). Among English-speaking children, SLI is identified on the basis of the clinical markers of the poor performance on nonword repetition tasks and the omission of finite verb morphology marking grammatical tense (e.g., Gathercole & Baddeley, 1990; Rice & Wexler, 1996; Tager-Flusberg & Cooper, 1999). We confirmed that children with autism who are impaired on standardized language tests perform poorly on nonword repetition tasks, showing the same error patterns that are reported for children with SLI. The children with autism also display difficulties on tasks designed to elicit the past tense (e.g., *She painted the wall*) and the third-person present tense (e.g., *She paints the wall*) and tend to omit specifically these morphemes in spontaneous speech. These kinds of deficits were not found for the children with autism in the "normal" language subtype.

This program of research on language in autism revealed that no single language phenotype defines autism. Rather, there are several distinct phenotypes, which include children with normal linguistic (but not pragmatic) skills and children whose phenotype is the same as SLI. These findings could only emerge from studies of autism that take a within-group individual difference approach that does not attempt to constrain the inclusion of participants to those who can be well-matched to the comparison participants. Of course, follow-up studies are warranted that would directly compare the "impaired" subtype to a group of children with SLI on a range of relevant language tasks. These groups would need to be well-matched on age and IQ in order to test whether these populations truly have the same language deficits.

Within the field of autism there is growing appreciation of the importance of studying the heterogeneity that is found across the spectrum of this disorder in all aspects of cognition as well as language (Dawson *et al.,* 2002; Happé, 2003; Tager-Flusberg & Joseph, 2003). As illustrated here, by directly investigating heterogeneity we can identify more homogenous subtypes within the population. In turn, these homogenous subtypes can potentially advance our understanding of the genetic and neurobiological bases of autism. For example, in the search for genes that confer susceptibility for autism, language subtypes were explored in two genetic analyses of multiplex families (Alarcon, Cantor, Liu, Gilliam, Geschwind, AGRE, 2002; CLSA, 2001). In one study, the subgroup of probands with autism who had no language or clearly impaired language whose parents also had a history of language difficulties were separated from the full sample that included children without language impairment (CLSA, 2001). The linkage signals on two chromosomes, 7q and 13q, both of which have been implicated as loci for SLI (Bartlett, Flax, Logue, Vieland, Bassett, Tallal, & Brzustowicz, 2002; O'Brien, Zhang, Nishimura, Tomblin, & Murray, 2003), were significantly increased, suggesting that these signals were mainly attributable to the language-impaired subtype within autism. Similar findings were obtained in the second study by the Autism Genetic Resource Exchange Consortium (AGRE) using different definitions of language-impaired in an independent sample of autism families (Alarcon *et al.,* 2002). These genetic findings hold out some promise that defining language phenotypic subtypes within the autism population may provide important benefits to genetic studies.

FUTURE DIRECTIONS FOR RESEARCH ON LANGUAGE IN AUTISM

Despite over five decades of research, there is still a great deal that needs to be learned about language in autism. There are many important issues that have yet to be investigated, and as illustrated here, no single methodological approach can be used to address different types of questions. For each research design, we need to be mindful of its advantages and disadvantages and pay attention to whether we have sufficient statistical power to answer our specific research questions. Matched group designs have their place, but investigators need to avoid the kinds of problems identified earlier.

Little is known about the *development* of language in autism. There have been few longitudinal studies conducted during the critical period when children with autism are in the process of acquiring language and none in which growth modeling techniques are used to investigate differences in the rates of development across language domains, or the kinds of growth patterns that may be found among children with autism. Such longitudinal studies can encompass both an individual difference approach and a comparative approach to explore variation in the development of language among children with autism (including when children begin to speak, growth patterns within and across different components of language, developmental rates and end points) and how different children with autism compare to other populations, such as SLI, in patterns of language acquisition. These kinds of studies are crucial for a more complete understanding of language and communication in autism that will inform basic research on underlying etiology and will ultimately lead to the development of new treatments that can improve the lives of children with autism, all of whom have significant deficits in this domain.

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